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The case for signs of joint hypermobility on disarticulated human bones

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Abstract: *Extended articular surfaces together with supra-condylar imprints at joints are identified as being extensions of the range of a joint movement in response to specific activity undertaken from an early age. They enhance and control the range of joint movement. Rarely, on skeletons recovered from archaeological contexts, an extra-large extended articulation indicates the limit of an extreme range where two bones of the joint made contact and arrested the movement of one over another. They are rarely observed even where stress activities were widely practised. It is suggested that these individuals had a pre-disposing physique—joint hypermobility. Hypermobility syndromes cannot be diagnosed on disarticulated skeletal remains since the full extent of joint mobility cannot be assessed, making standard diagnostic criteria inapplicable. If, however, extra-large extended articulations and bone imprints can be considered as additional signs of joint hypermobility, that assessments of joint hypermobility from historical samples may be feasible.*

Key words: joint hypermobility syndrome; Neolithic; extended joint articulation; bone imprint; stop facets

Introduction

Habitual constrained activities from childhood can result in the development of extended articular facets of joints of the spine, hips, knees, ankles or toes. Extended articular facets are normal morphologies often enhanced by a range of specialist tasks that can be characterised by modifications to specific joints. When associated with activity related positions, extended or modified articular joint surfaces can develop on cervical vertebrae as strong uniform processes stabilizing the head, on thoracic vertebrae as buttressed vertebral articulations stabilizing the spine in porters using backpacks, or at the femoral neck of horse riders (Ubelaker 1979; Kennedy 1989; Sofaer 2000; Mann et al. 2016; Molleson 2016). These modified joint surfaces are best seen on skeletal material. They can be particularly impressive on archaeological skeletons dating from times when individuals spent many hours in repetitive activity starting

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at an early age while still growing. Exceptionally, extra-large articulations have been observed on joints of the spine, elbow, fingers and thumb, ankle and foot.

From Neolithic times, when goods had to be transported over moderate distances, a bag might be slung over the shoulder. Although much of the time the bag would have been heavy enough to force the head of the radius onto the humerus, only infrequently would an imprint of the head of the radius develop on the ventral shaft lateral to the ulnar sulcus (**Figure 1**). A radial imprint lateral to the ulnar sulcus was noted on a humerus from House 8A at Neolithic Nemrik, Iraq (Molleson 2006, Fig. 4). The thumb of Tr.B73.B66 from Tell Abu Hureyra PPNB (Phase 8) has ventral (palmar) supracondylar imprints on both distal and proximal phalanges. They appear to be insertion pits of *flexor pollicis longus*. There is a similar type of pit, perhaps for *extensor pollicis longus*, on the dorsal of the distal end of the proximal phalanx. A supracondylar bone imprint can develop on the shaft of the first metatarsal proximal to an extended articular facet where the proximal phalanx of the big toe impacted during kneeling with the weight bearing on the toes. Bone imprints can also develop at the ankle arresting the impact of the talus on the tibia, where further flexion of the ankle would normally be restricted by the Achilles tendon. If this tendon is lax, the movement continues until the talus is stopped by impact with the tibia and a supracondylar bone imprint develops as a result. Such imprints are rare even in populations where activity driven kneeling is widely practised. They can be continuous with or detached from an extended articular surface. Such imprints are paradoxical in that they form as a stop to further movement where one bone of a joint impacts on the other, thus stabilizing the joint by arresting excessive movement around the joint surfaces. Like an extended or modified articular facet, an accessory facet, or a buttressed facet, they act to stabilize a joint (Jones & Holbrook 2016).

The sporadic occurrence of hyperflexed or hyperextended joints among groups of craft specialists, in which the morphologies are generally more moderate, raises the question that some at least occur in those who had particularly mobile joints. The signs of joint hypermobility, which is diagnosed if a specific joint extends over more than 90° and includes manifestations of various Herited Disorders of Connective Tissues (HDCT) syndromes, including benign joint hypermobility, Ehlers-Danlos syndrome and Marfan habitus (Grahame 2003; Malfait et al. 2017). The generally low incidence (5%-11%) of a joint hypermobility syndrome (JHS) in most Western populations would explain the rarity of extremely large articular facets and supracondylar imprints even where kneeling to use a saddle quern or mortar, for example, was a normal way of preparing grain for cooking. Rodney Grahame (1986) found only 2% of 9275 patients attending a rheumatology clinic were diagnosed with JHS and of these 85% were female (Keer & Grahame 2003:71). Much higher frequencies have been recorded in some African and south west Asian populations: 25.4% in

Table 1. Morphological signs on bone that might indicate joint hypermobility. The Beighton score is an edited version of the Carter/Wilkinson scoring system which was used for many years as an indicator of widespread hypermobility. Medical professionals varied in their interpretations of the results; some accepting as low as 1/9 and some 4/9 as a diagnosis of hypermobility syndrome. Therefore, it was incorporated, with clearer guidelines, into the Beighton criteria. The Beighton score is measured by adding 1 point for each of the following.

Beighton sign	Possible skeletal sign
Placing hands flat on the floor with straight legs	Imprint on anterior femoral neck
Left knee bending backward Right knee bending backward	Curved anterior rim of distal articulation of femur and proximal dorso-lateral articulation of tibia
Left elbow bending backward Right elbow bending backward	Deep imprint of radius head on distal end of humerus shaft
Left thumb touching the forearm Right thumb touching the forearm	Modification of proximal articulation of trapezium on medial rim of distal articulation of radius
Left little finger bending backward past 90° Right little finger bending backward past 90°	Dorsal extension of distal articulation of 5th metacarpal

males and 38.5% in females among 1774 students from Iraq (Al-Rawi et al. 1985); 18% in girls and 14.4% in boys among 997 Egyptian children (El-Garf et al. 1998) and 35% in boys and 57% girls in a sample of 204 West African children (Birrell et al. 1994). Wordsworth and colleagues in an early study of regional variation found Asian Indians to be significantly more mobile than English but less so than Iraqi students (Wordsworth et al. 1987; Keer & Grahame 2003:71; McKusick 1960:169).

JHS feature joints (especially the matacarpal-proximal phalangeal joint of the fifth finger) that easily move beyond the normal range. Rare and variable in its presentation in living populations JHS is difficult to diagnose and several criteria are used, notably the Beighton score system based on the Beighton criteria (Beighton et al. 1973; Keer & Grahame 2003; McKusick 1960; Neki & Chhabra 2016; Remvig et al. 2007; Simpson 2006; Tinkle et al. 2017). Criteria include hyper-extended (greater than 90°) phalangeal joints of the thumb and metacarpo-phalangeal joints, proximal and distal phalangeal joints of the index finger. The Beighton scores assess joint hypermobility syndrome as being present when 4 or more of 9 tests are positive. None of these tests, unfortunately, are applicable to disarticulated skeletal material. These are summarised on **Table 1**. HDCT essentially concern soft tissues such as tendons to joint muscles (**Figure 1**). Joint hypermobility is more frequently diagnosed in females than males.

Rodney Grahame has argued that the search for hypermobile joints should look beyond the few that constitute the Beighton score (Grahame 2003:5, Box 1.3, Fig. 1.2; Grahame and Hakim 2013). As well as modelling of joint articular surfaces dental crowding with high narrow palate; arachnodactyly, measured on the metacarpals;

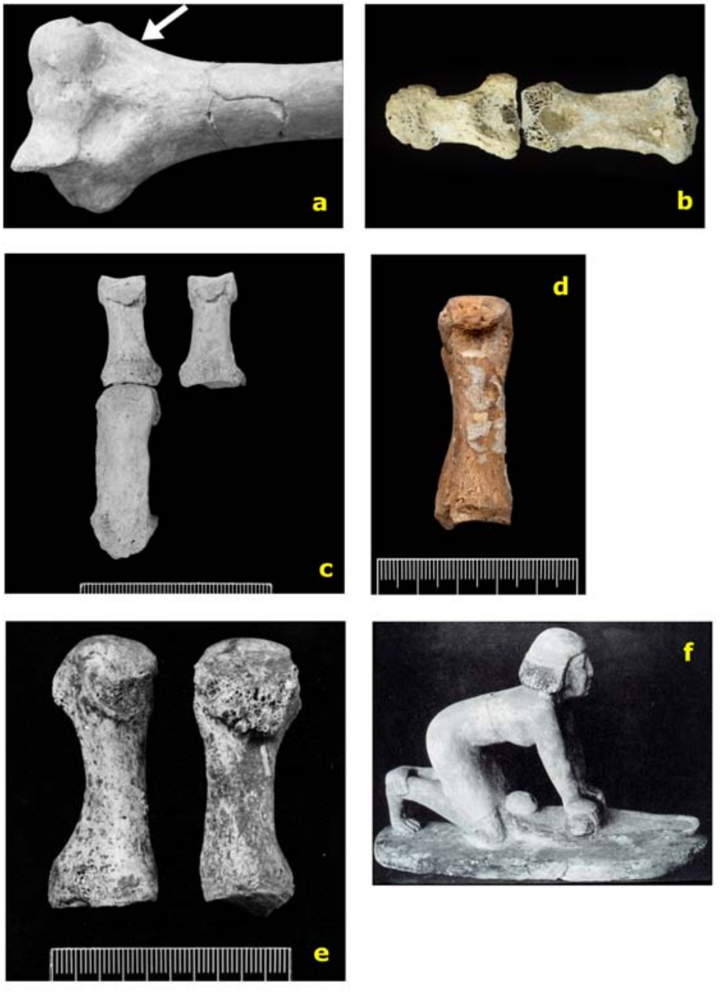

Figure 1. Imprints. a) Shallow imprint of radius bone lateral to ulnar sulcus on supracondylar region of distal humerus, denoting limitation of flexion of forearm. House 8A Neolithic Nemrik, Iraq (H8Ask3402 Archaeological Museum Warsaw, Poland); b) Dorsal imprint prossibly of *extensor pollicis brevis* on proximal phalanx of thumb; and anterior (palmar) imprints on both terminal and proximal phalanges of flexors of gripping thumb of the same individual. Neolithic Tell Abu Hureyra, Syria Tr.B73.B66 Ph8 Rm.3 (NHMUK); c) Hyperextended thumb articulations: dorsal view of right and left proximal phalanges with ossified tendons of extensors on proximal phalanges and medial view of proximal phalanx of thumb and first metacarpal (MCI). Male, Modern Era Tell Abu Hureyra, Syria Tr.E72.178 (NHMUK); d) Imprint of extensor proximal to extended articulation (kneeling facet) of first metatarsal. Adult female Neolithic Tell Abu Hureyra, Tr.C72.553A (NHMUK); e) Bone imprint on right first metatarsal (MTI) and traumatic osteoarthritic lipping on left MTI. Adult female, Neolithic Tell Abu Hureyra Tr.Tr.B73.2949 (NHMUK); f) Egyptian figurine using a saddle quern rests the left foot on strongly flexed right ankle (from Breasted 1913).

stature and arm span to height ratio >1.05. But arm span cannot be estimated on skeletal material. Here it is proposed that certain extreme articulations which are occasionally observed on disarticulated skeletal bones could be indicative of JHS.

Not all with hypermobile syndrome manifest extreme joint morphology although most will have slender bones especially of the limbs, hand and foot bones. Arachnodactyly, as defined by the Metacarpal Index >8.6, is one of the few signs of Erlers-Danlos Syndrome (EDS) that can be applied to disarticulated skeletal material. The Metacarpal Index is the mean mid-length to width ratio of the four metacarpal bones, omitting the thumb. Dolichocephaly (100W/L <75) and high arched palate with dental crowding can only occasionally be measured on excavated material, which is usually fragmented. There are single measures from Tell Abu Hureyra, Syria, for the Neolithic, 72.1, which is in the dolicocephalic range and one for the Modern Era, 75.14, mesocephalic (Molleson in Moore et al. 2000).

Extremely large articular facets and supra-condylar imprints occasionally observed on disarticulated skeletal material would have formed in response to extreme extension or flexion of the joint. Often the full range of movement can be inferred to have been greater than 180°; but it is not known to what extent they match up with any of the many forms of hypermobility. An examination of the range of signs from the Eastern Mediterranean region, where high frequencies of hypermobility have been recorded in the living, might possibly enhance the diagnostic criteria used to define hypermobility. Two major skeletal collections Tell Abu Hureyra, Syria, and Çatalhöyük, Turkey, which had already been described, were re-assessed for possible signs of JHS (Moore et al. 2000; Hodder 2005). A few additional cases from other sites in the region are also included. They span major cultural levels from the Neolithic to the Modern Era times. The earliest have been noted on hand and foot bones from Neolithic times, 10.000-8.000 years ago (**Table 2**).

Evidence from the Near East

Two fairly complete skeletons, one Neolithic from Çatalhöyük, the other from Modern Era contexts from Tell Abu Hureyra, illustrate some of the signs of JHS inferred on skeletal material (**Figures 2** and **3**). Other examples are from very incomplete skeletons on which an extended range of movement is evident in the joint morphology although the full extent of hypermobility cannot be assessed (**Figure 1**).

CH96.Sk1378 represents the crouch burial of an edentulous old man from Neolithic Çatalhöyük, Turkey (**Figure 2**). He was taller, 1.713m (5ft 7in), than the six other males (1.543-1.694m) for whom stature could be estimated from long bone length. The old man had open cranial sutures (Molleson et al. 2005, Figs 12.1, 12.6, 12.7). The metacarpal index, MI=7.38, does not specify arachnodactyly. He has ossified extensions of the articular surfaces of thumb proximal phalanges. These appear

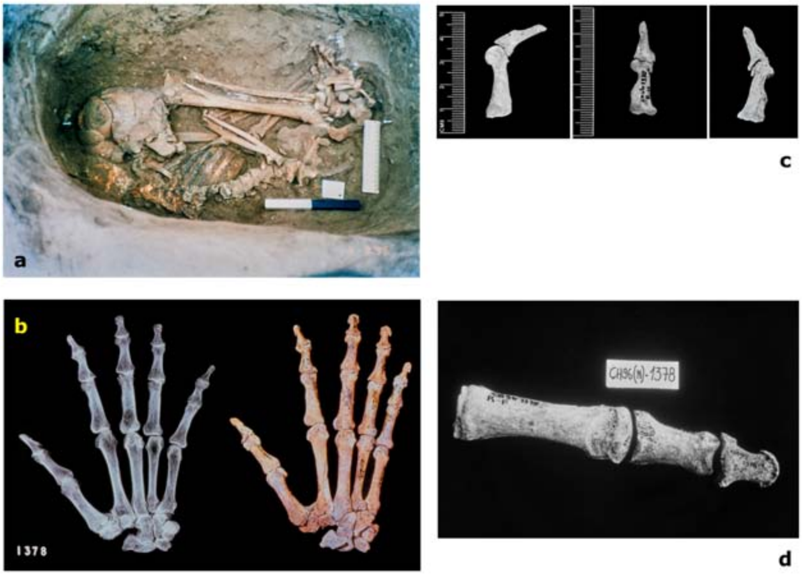

Figure 2. Signs of hypermobility on joints of disarticulated skeleton of old male from Çatalhöyük, Turkey: a) crouched burial of CH96.Sk1378 in situ; b) dorsal and radiographic views of reassembled hand bones; c) phalanges of thumb, index and fifth fingers; d) bones of big toe with imprint of proximal phalanx on first metatarsal. Photos: Photo Studio NHM; Genis Ribe, Çatalhöyük excavations.

to be ossified insertions of *extensor pollicis longus* tendons. Extension of the distal phalanx would have extended over more than ninety degrees; the distal articulation of the middle phalanx of the fifth digit is also extended (while that of the index is deformed). A similar development is seen on the proximal phalanges of the thumbs of a male, Tr.E72.178, from Modern Era Tell Abu Hureyra (**Figure 1**).

Articular surfaces of the first metatarsal of CH96.Sk1378 also have a supra-condylar imprint where the proximal phalanx of the big toe impinged on the metatarsal during habitual kneeling or squatting on his toes. Soot deposits, visible on his ribs in the excavation, indicate that he may have been house-bound, developing anthracosis in the smoky atmosphere of the house, then hyperparathyroidism, which was recognized on radiographs of the hand bones by Basil Shepstone at The Radcliffe Hospital, Oxford (Molleson 2007). A long term aspect of chronic disease might predispose to generalized joint laxity (Grahame 2003:2).

The MTI of Tr.B73.2949 from Tell Abu Hureyra is another individual who has an extended articulation of the big toe and shows in addition the osteoarthritic consequences of trauma sustained perhaps through over-shooting the saddle quern by too energetic grinding (**Figure 1**).

A female Tr.A72.338D from Modern Era Tell Abu Hureyra, Syria, displays several possible signs of hypermobile joints in the exceptionally marked responses to intense activity. The tallest woman at 1.65m (5ft 5in) of the sample (1.43-1.65, mean 1.55m) she had very large bone imprints on both tibia and talus limiting hyperflexion of the ankle. There is bony lipping of the head of the first metatarsal (MTI) indicating that the big toe joint could have extended over at least 90 degrees. These signs could be associated with a pounding or grinding activity from a kneeling position. She also had unusually pronounced pits in the pre-auricular sulcus of the ilium (**Figure 3**).

Extremely deep pits of ligament attachment in the pre-auricular sulcus, known as 'scars of parturition', were also noted on Tr.A73.B44 and Tr.C73.77A, both from Modern Era Tell Abu Hureyra. Increased laxity of the ligaments due to hormonal changes during pregnancy with increased mobility of the pelvic joints may be responsible for these; an increase in collagen laxity would result in less ability to resist

Table 2. Some possible signs of joint hypermobility in archaeological skeletons.

Culture\Site	ID	Bone	Sign
		Neolithic	
Iraq, Nemrik	H8Ask3402	humerus	supracondylar radial imprint
Syria, Abu Hureyra	Tr.B73.B66	thumb phalanges	hyperflexed, hyperextended pits
	Tr.B72.2949	MTI	bone imprint left; arthritis right
	Tr.B72.2950	MTI–big toe	bone imprint right
	Tr.C72.553A	MTI	kneeling facet, bone imprint
	Tr.C72.389A	tibia	squatting facet
Turkey, Çatalhöyük	CH96.Sk1378	cranium	open sutures
		hand; thumb	long MCI-IV; extended facets
		scapula, ribs	brown tumour, unhealed fractures
		MTI; ankle	kneeling facet; bone imprints
		Bronze Age	
Iraq, Ur	G154.AH	sacrum	accessory dorsal facets
		ilium	'scars of parturition'
	PG 1648d	sacrum	accessory dorsal facets
		Modern Era (Historical)	
Syria, Abu Hureyra	Tr.A72.338D	humerus	radius imprint
		ankle, MTI	tibio-talus imprint; kneeling facet
		ilium	'scars of parturition'
		scapula; rib	acromion detached facet; bifid rib
	Tr.A72.102	humerus	radius imprint
	Tr.A73.B44	ilium	spondylolithesis L4,5; 'scars of parturition'
	Tr.A72.275B	MTI–big toe	kneeling facets, bone imprints
	Tr.C73.77A	humerus	radius imprint; birthing scars
	Tr.C72.105	lumbar vertebrae	spondylolisthesis
	Tr.E72.178	thumb phalanges	hyperextended prox. phalanges

Figure 3. Possible signs of joint hypermobility displayed by one individual: a) anterior bone imprints on left tibia and left and right talus bones indicate hyperflexion of both ankle joints; b) extended articulation on first metatarsal (MTI) with osteoarthritic lipping; c) unusually large deep pits ('scars of parturition') in the pre-auricular sulcus of the ilium (not to scale). Old adult female, Modern Era, Tell Abu Hureyra, Tr.A72.338D (NHMUK).

stretching (Ostgaard et al. 1993; Keer & Grahame 2003:81). Al-Rawi et al. (1985) noted that, in general, ligament sprains were reported by Iraqi students. Ligamentous rupture, the association of ligamentous rupture with genital prolapse in women, especially multiparous women, is part of generalized connective tissue disorder (Al-Rawi & Al-Rawi 1982). In general few females of child bearing age show marked signs of parturition, which raises the possibility that an individual's underlying physique could influence the development of deep ligament scars as much as the position taken up during birthing: a seated position imposing less strain on the pelvis than lying supine. Even the hypermobile joint will be susceptible to injury if the joint is driven beyond the limits of any holding ligaments that may be strained. One of the women, Tr.A73.B44, had spondylolithesis of the 4^{th} and 5^{th} lumbar vertebrae and had a detached facet on the left acromium. She must have sustained a right shoulder injury while still growing—the sternum has an expanded area of attachment for the *pec-*

toralis major, like that of the Jericho cripple (Molleson 2016). She was probably quite disabled, moving about with a stick-crutch.

Discussion

In this overview of disarticulated skeletal material from two major Near Eastern sites two related signs have been identified as possibly being indicative of joint hypermobile syndrome. Extremely extended articular joint facets and supracondylar imprints both reflect a greater than 90° range of joint flexion or extension. Anomalous ligament scars of the pelvis and failure of the neural arch of a vertebra to unite can be associated with the syndrome (Keer & Grahame 2003:55). It is impossible to estimate incidence of extreme facets and imprints in archaeological samples given the variety of activities and variation in practices. Supracondylar imprints appear to be the best indicators of extreme joint range. Since the incidence of JHSs in Europeans is around 5-25% it is unsurprising that JHS is largely unknown even today. Another major factor is the effect of evolving changes in production techniques: the saddle quern is no longer in general use and load bearing tasks have greatly increased. These changes in techniques may have alleviated the effects of JHS in those who do not tolerate excessive or repetitive tasks.

The earliest extended facets have been recorded on Neolithic bones when intensive skills-defined roles are likely to have been more widespead than the few cases identified. The thumb bones from Neolithic Tell Abu Hureyra, Tr.B73.B66 (**Figure 1**), were recovered from Room 3 (Phase 8) which has been identified as the place where a different group, newly arrived and possibly from Africa, buried their dead (Molleson & Rosas 2012). The mature male Tr.C73.845 with artificially deformed asymmetric skull and noticeably gracile limbs, dates from this time (Molleson 2016, Fig. 6). He has strong unciate processes of a head-loader or porter. At present the respective impacts of acquired behaviour and inherent physique are not clear. None of those with signs of joint hypermobility could be shown, from other non-metric traits, to be related.

Obesity, where the joints are involved, would limit joint flexion but usually only develops later in adulthood after bone development is complete, whereas extended facets and bone or tendon imprints must start to develop during growth. There is a joint-protective effect of hypermobility. The distinctive slender build of many individuals with JHS could be advantageous in hot climates, while cold temperatures have a negative impact (Wells & Cole 2002; Mangharam 2003:135). Osteoarthritis of joint articular surfaces would be an incidental consequence of repetitive stress sustained during strenuous activities, particularly when taken up in adulthood, but would not be part of a syndrome. Kraus et al. (2004) argued that there was no evidence for increased odds of osteoarthritis of the hand in any joint group in association with articular hypermobility.

This is very much a preliminary attempt to highlight the possibility that a variety of enlarged articulations, facets and imprints could be considered as signs of joint hypermobility syndrome in the context of disarticulated skeletons. From Neolithic times very young children would have been incorporated into the intensive repetitive labour activities of farming and food preparation. This was their schooling. Specific indicators of many such activities, such as squatting facets, have been documented in skeletal material. It is suggested that the extreme development of some of these extreme and paradoxical facets and stabilising stop imprints can be highlighted as signs of a joint hypermobility syndrome. A key additional observation would be the shape of the hard palate (Burris & Harris 2000), not routinely measured by anthropologists. A number of recent orthodontic studies could be the basis of a reference data base (Sakuma et al. 2009; Thiyagarajan 2008; Yang et al. 2013).

Conclusions

While most of the standard criteria of a joint hypermobility syndrome cannot be defined on disarticulated skeletons, both extra-large facets and bone or tendon imprints might be considered signs where they indicate hyperflexion or hyperextension of a joint, even where paradoxically they limit articulation. Strikingly pronounced articular facets or supracondylar imprints on individual bones could be indicative signs of lax joints. They are associated with activity related skeletal morphologies and notably date from Neolithic times with the emergence of labour intensive repetitive skills.

Despite the limitations of disarticulated skeletal material it seems that there is evidence for joint hypermobility in the past and some kind of a history and distribution of these rare and highly complex variants of the human genome could be recorded. At least, if extreme facets and imprints can be considered, it is possible to survey for these signs and by taking into account the known diagnostic signs so enhance understanding of the complexities, aetiologies and origins of joint hypermobility.

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